CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 5 (2014) 274–276

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Adenomatoid hyperplasia of the minor salivary glands on the buccal mucosa: A rare case report

Ömür Dereci[a,*], Emre Çimen[b]

[a] Eskişehir Osmangazi University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Eskişehir, Turkey
[b] Oral and Maxillofacial Surgeon, Private Practice, İstanbul, Turkey

ARTICLE INFO

Article history:
Received 3 February 2014
Accepted 19 March 2014
Available online 25 March 2014

Keywords:
Adenomatoid hyperplasia of the minor salivary glands
Minor salivary glands
Adenomatoid hyperplasia
Buccal

ABSTRACT

INTRODUCTION: Adenomatoid hyperplasia of the minor salivary glands is a hyperplastic oral lesion which may be seen on minor salivary gland bearing areas on all oral mucosa, especially on soft and hard palate. This study reports a rare case of buccal adenomatoid hyperplasia of the minor salivary glands and discusses the clinical significance.
PRESENTATION OF CASE: 48 year old male patient presented with a complaint of a swelling on his left cheek. Clinical examination revealed a bluish mass on the buccal mucosa. A provisional diagnosis of salivary gland neoplasm was made and the lesion was excised under local anesthesia. The histological diagnosis was adenomatoid hyperplasia of the minor salivary glands.
DISCUSSION: Buccal localization of the adenomatoid hyperplasia of the minor salivary glands is quite uncommon in the literature. There are two cases in the English literature for our knowledge. In the clinical examination, the nodular and protuberated appearance of the lesion resembles buccal minor salivary gland tumors and vascular lesions. Histological analysis is fundamental to achieve correct diagnosis.
CONCLUSION: The differential diagnosis of buccal nodular, exophytic and colored mucosal lesions should include adenomatoid hyperplasia of the minor salivary glands.

1. Introduction

Adenomatoid hyperplasia of the minor salivary glands is a very rare, hyperplastic lesion that is first described by Giansanti et al. in 1971.[1] It is an asymptomatic tumor-like mass with localized hyperplasia of the minor salivary glands. It is associated with localized mucosal swelling and clinically imitates a salivary gland neoplasm.[2,3] Microscopically, lesion contains normal – appearing salivary gland tissue.[2]

Etiology of the lesion is unknown. It is suggested that adenomatoid hyperplasia is not associated with specific factors (drugs, nutritional deficits, anorexia nervosa, neurogenic factors) which may cause sialodenithis or enlarged major salivary glands.[2]

This study reports a case of adenomatoid hyperplasia of the minor salivary glands on the buccal mucosa which is a rare localization for this entity. There are only 2 reported cases of buccal localization of the lesion in the literature.[2] And it is thought to be the first literature reporting a buccal localization of the lesion in the Turkish population.

2. Presentation of case

A 48 years old non-smoker male patient presented with a complaint of swelling at his left buccal mucosa. Clinically, the lesion had red-bluish color, exophytic nodular appearance and smooth and soft texture (Figs. 1 and 2). There was no pain or tenderness. No regional lymphadenopathy was observed. The medical history of the patient was non contributory. Also there were no signs and history of trauma at the site of the lesion. Panaromic radiography showed no abnormal findings. It was suspected that the lesion might be a vascular lesion, however, no pulsation was recorded with bidigital palpation and diascopy test was negative. Then, the provisional clinical diagnosis was made as salivary gland neoplasm. The lesion was excised under local anesthesia (Articain HCL 80 mg, epinephrin 0.020). Postoperative healing was uneventful.

2.1. Histological findings

Microscopically, surgical specimen sections stained with hematoxylin and eosin. Epithelial layer of the mucosa showed acanthosis, edema and verrucous hyperplasia. Edematous connective tissue and prominent vascular proliferation were observed on the subepithelial region. The maturation of the epithelium was normal and no dysplasia was detected. In the submucosal layer, there were minor salivary gland tissues containing hyperplastic nodular mucous

* Corresponding author at: Eskişehir Osmangazi University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, 26480 Meşelik Campus/Eskişehir, Turkey. Tel.: +90 2222391303; fax: +90 2222391273.
E-mail address: omurdereci@hotmail.com (Ö. Dereci).

http://dx.doi.org/10.1016/j.ijscr.2014.03.020

Fig. 1. Intraoral view of the buccal adenomatoid hyperplasia.

Fig. 2. The lesion is protuberated from the buccal mucosa masquerading a salivary gland neoplasm.

acini.(Fig. 3) Also, some minor salivary gland nodules could be seen in the muscular tissue. There were no signs of neoplastic transformation

3. Discussion

Adenomatoid hyperplasia of the minor salivary glands is a localized hyperplastic nodular mass appearing as soft or firm swelling on the oral mucosa.[2,4] The color of the lesion is mostly in the color of normal mucosa but it is reported that it might be slightly reddish.[2] In our case the lesion was bluish red. Generally the lesion is asymptomatic. Adenomatoid hyperplasia is predominantly seen in males.[4] There is no age predilection.[2,4,5]

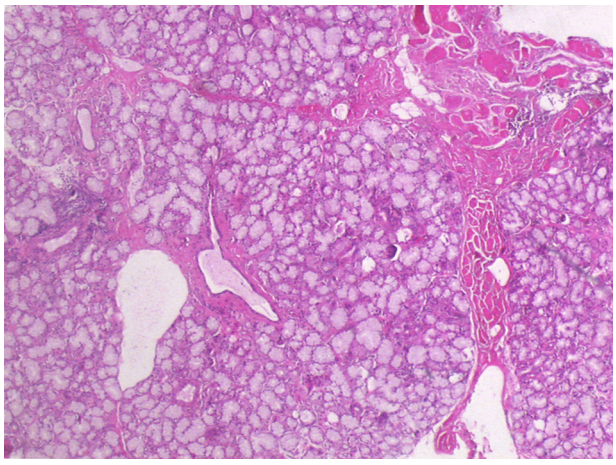

Fig. 3. Histological section of the lesion reveals hyperplastic minor salivary gland tissue with prominent mucous component. There is also dispersed ectopic minor salivary gland tissue in the skeletal muscle. (HEX100).

The differential diagnosis of the lesion must include benign and malignant salivary glands neoplasms such as mucoepidermoid carcinoma, adeno cystic carcinoma and pleomorphic adenoma.[3,4] Since the lesion has no specific clinical appearance, the certain diagnosis should be done microscopically.[2]

Histopathologically, the microscopical view of the lesion contains multiple clusters of normal appearing mucous acini surrounded by fibrous connective tissue. There is an increase in the number of acini. Inflammatory reaction is rarely seen in the lesion.

There are several reported cases of adenomatoid hyperplasia of the minor salivary glands. Only 3 large series have been reported by Arafat et al., Buchner et al. and Barret et al. with 10, 40 and 20 cases respectively.[2,4,5] Approximately 90 cases have been reported in the English literature so far.[2,5–8] Palatal mucosa is the most common localization for the lesion. It can be found both in soft and hard palate. Other sites such as retromolar area, lip and mouth floor are also seen to be affected.[2,5,8,9] Buccal mucosa localization was only reported for 2 cases in all English literature.[2] The reason of the rarity of buccal localization may be explained with the low amount of the minor salivary glands in the buccal region compared to the palate and other parts of the oral mucosa.

The treatment of the lesion is total excision.[1,2] No reccurence has been reported in the literature yet. However, it is reported that one palatal case has developed into mucoepidermoid carcinoma several years after diagnosis.[5] Malignant transformation possibility of the lesion was investigated by cytogenetic analysis and translocation t(2;14)(q21;q22) have been found in the lesion.[10] However, it is not known whether this chromosomal aberration may be a risk for malignancy.

4. Conclusion

Adenomatoid hyperplasia of minor salivary glands is a rare, benign hyperplastic lesion which is localized mostly on palatal regions. Buccal localization, as in this case, is quite uncommon and may be confused with benign or malign salivary gland tumors and vascular lesions clinically. The differential diagnosis of protuberated, colored and nodular lesions on the buccal mucosa should include adenomatoid hyperlasia of the minor salivary glands.

Conflict of interest

None declared.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

Author contributions

Ömür Dereci: Design, data Collection, writing the paper. Emre Çimen: Data collection, writing the paper.

Key learning points

The differential diagnosis of buccal nodular, protuberated and colored mucosal lesions should include adenomatoid hyperplasia of the minor salivary glands.

References

1. Giansanti JS, Baker GO, Waldron CA. Intraoral, mucinous, minor salivary gland lesions presenting clinically as tumors. *Oral Surg Oral Med Oral Pathol* 1971;**32**:918–22.
2. Buchner A, Merrel PW, Carpenter WM, Leider AS. Adenomatoid hyperplasia of minor salivary glands. *Oral Surg Oral Med Oral Pathol* 1991;**71**:583–7.
3. Scully C, Eveson JW, Richards A. Adenomatoid hyperplasia in the palate: another sheep in wolf's clothing. *Br Dent J* 1992;**173**:141–2.
4. BarretAW, Speight PM. Adenomatoid hyperplasia of oral minor salivary glands. *Oral Surg Oral Med Oral Pathol* 1995;**79**:482–7.
5. Arafat A, Brannon RB, Ellis GL. Adenomatoid hyperplasia of mucous salivary glands. *Oral Surg Oral Med Oral Pathol* 1981;**52**:5.
6. Brown FH, Houston GD, Lubow RM, Sagan MA. Adenomatoid hyperplasia of mucous salivary glands: report of two cases. *J Periodontol* 1987;**58**:125–7.
7. PetriWH, Carr RF, Kahn CS. Adenomatoid hyperplasia of the plate. *J Oral Maxillofac Surg* 1993;**51**:310–1.
8. Barret AW, Speight PM. Adenomatoid hyperplasia of oral minor salivary glands. *Oral Surg Oral Med Oral Pathol* 1995;**79**:482–7.
9. Chen YK, Lin CC, Lin LM, Yan YH. Adenomatoid hyperplasia in the mandibular retromolar area: case report. *Aust Dent J* 1999;**44**:135–6.
10. Manor E, Sinelnikov I, Brennan PA, Bodner L. Chromosomal aberrations in adenomatoid hyperplasia of palatal minor salivary gland. *Br J Oral Maxillofac Surg* 2013;**51**:170–2.